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Case report

Successful bronchoscopy in a pregnant patient with plastic bronchitis

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ABSTRACT

Plastic bronchitis is a rare disorder, characterized by formation of thick fibrinous bronchial casts which can obstruct the airway and present as a life threatening emergency (1). It is more common in the pediatric population after corrective or palliative surgery for congenital heart disease like fontan procedure but has rarely been reported in adults as well (1). Pregnancy is a relative contraindication for bronchoscopy. Bronchoscopy in the pregnant patient poses significant risks as manipulation of the airway can lead to impaired oxygenation and ventilation. In addition, the drugs used during this procedure to provide sedation can have a direct impact on the developing fetus (2). In spite of these risks bronchoscopy should not be withheld in an emergent situation as it can be a lifesaving measure. We report a case of successful bronchoscopy using Propofol as the sedating agent in a pregnant female with plastic bronchitis who presented with respiratory distress.

1. Case description

A 19 year old female presented to the emergency room with complaints of cough associated with thick gelatinous expectoration and dyspnea that had started 6–8 hours prior to presentation. She had a history of congenital heart disease for which she had undergone Fontan procedure in her infancy. Prior to this episode, she had multiple similar episodes and was diagnosed with plastic bronchitis. Her vitals were initially stable but then she developed hypoxia with oxygen saturation dropping to 90% requiring 100% FiO2. On her initial blood work she was found to be pregnant. She was in her first trimester and was G1P0L0 at 12 weeks of gestation. No radiologic images were obtained. Given her history of plastic bronchitis and her progressive respiratory distress, bronchoscopy was deemed necessary for both diagnostic and therapeutic purposes. Potential risks to the mother and fetus and potential benefits were explained and informed consent was obtained. Bronchoscopy was immediately performed after intubation and low dose Propofol infusion (0.025 mg/kg/min) was used for sedation. Given the increased maternal and fetal risk with sedatives, Propofol was chosen (class B drug) and fentanyl intravenous boluses of 50 mcg (Class C) were used as needed. She received a total of 175 mcg of fentanyl. On inspection, there was a large plaque like secretion completely obstructing the left main bronchus extending beyond the carina into the trachea. Using forceps, a large bronchial cast was extracted from trachea and left main bronchus as shown in Figs. 1 And 2. Inspection of the rest of the airways showed otherwise patent airway. Hypoxia improved after removal of casts but patient remained intubated for a period of 48 hours. She became hypoxic on day 2, bronchoscopy was repeated to remove residual cast. During the second bronchoscopy the cast had recurred in the left main bronchus, this time endobronchial N-acetylcystine was used as an adjunct to help remove the cast. Patient tolerated the procedures well. The patient had no further exacerbations from plastic bronchitis during the rest of the pregnancy and had a successfully delivery at 37 weeks. The microscopic evaluation of the bronchial cast showed acellular material and bronchial lavage was negative for infection.

2. Discussion

Plastic bronchitis is a rare disorder characterized by formation of casts of varying size that can cause obstruction of an entire lung [3]. Bronchial casts can be seen in diseases associated with diffuse bronchial hypersecretion like asthma, bronchopulmonary aspergillosis, mucoviscoidosis [4–6]. They can also occur in patients with congenital heart disease particularly after surgical correction using the Fontan procedure where it can occur in up to 4% of patients [1].

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Fig. 1. The macroscopic appearance of the bronchial cast.

The exact pathophysiology of plastic bronchitis in congenital heart disease is unknown. Lymphatic dysfunction, endobronchial lymph leakage, and mucus hypersecretion due to elevated venous pressure have all been proposed as possible mechanisms [7]. Plastic bronchitis has been classified based on histology into cellular and acellular [8]. Alternatively, plastic bronchitis has been classified according to the associated disease state [9]. The cast morphology of plastic bronchitis in patients with congenital heart disease post corrective surgery is typically acellular and presents as a recurrent disease, like our patient.

Plastic bronchitis can present as a life threatening condition as in our patient. Cardiopulmonary stabilization with intubation and mechanical ventilation is the main stay of treatment in acute life threatening emergencies. Airways clearance with immediate rigid or flexile bronchoscopy is often required in such conditions. The risk of bronchoscopy in a pregnant patient is unknown and the literature is limited to case reports and review articles [2]. There is

Fig. 2. The macroscopic appearance of the bronchial cast expectorated by the patient.

procedural risk, risk from medications due to altered physiology in the pregnant patient and potential teratogenicity to the growing fetus. Thus careful understanding of maternal-fetal interaction of these medications is necessary when choosing the sedative agent. Peripartum it can cause neonatal respiratory depression [10]. Our patient was in her first trimester of pregnancy, and given the acceptable safety profile of propofol (class B) compared with the usual alternative of benzodiazepine (class D), we chose propofol as our sedative agent.

Removal of a large bronchial cast has been successful using multiple endoscopic instruments (forceps, baskets or snares) and leads to significant improvement in respiratory status. Use of cryotherapy to remove a large bronchial cast en bloc has also been reported [11]. In our case, we were able to remove the cast using forceps. In addition, during the second bronchoscopy we used direct instillation of N-acetylcystine during the bronchoscopy to facilitate removal of cast. According to Brogan et al., acetylcystine has the greatest effect in dissolving these casts over DNase, urokinase and tPA [12]. Due to the complexity of the disease, prevention of recurrence of plastic bronchitis post Fontan procedure is very challenging. Use of inhaled mucolytic and systemic anti-inflammatory medication has been reported to potentially reduce recurrence [3]. Patients who undergo heart transplant due to progression of their underlying disease do not present with recurrence post-transplant. Our patient refused any chronic therapy due her pregnancy and she progressed to 37 weeks of gestation without recurrence and was successfully delivered at 37 weeks due to her high risk pregnancy state secondary to congenital heart disease.

In conclusion, plastic bronchitis is a rare disorder which can present as a life threatening emergency and its presence in conjunction with pregnancy can make management very difficult. Bronchoscopy which would be otherwise contraindicated in pregnancy can be entertained when it is required to treat a life threatening condition and there are no alternative therapies. As demonstrated in this case, with judicious use of appropriate agents, bronchoscopy can be successfully employed during pregnancy with good outcomes for both the mother and the fetus.

References

[1] G. Grutter, D. Di Carlo, F. Gandolfo, R. Adorisio, S. Alfieri, G. Michielon, et al., Plastic bronchitis after extracardiac Fontan operation, Ann. Thorac. Surg. 94 (2012) 860–864.

[2] I.J. Bahhady, A. Ernst, Risks of and recommendations for flexible bronchoscopy in pregnancy: a review, CHEST J. 126 (6) (1974-1981).

[3] Eberlein, H. Michael, Michael B. Drummond, Edward F. Haponik, Plastic bronchitis: a management challenge, Am. J. Med. Sci. 335 (2) (2008) 163–169.

[4] R.D. Fairshter, C.A. Riley, R.I. Hewlett, Large bronchial casts, Arch. Intern Med. 139 (1979) 522–525.

[5] A. Bowen, K. Qudjhane, K. Odagiri, S.L. Liston, W.A. Cumming, K.S. Oh, Plastic bronchitis: large, branching, mucoidbronchial casts in children, Am. J. Roentgenol. 144 (1985) 371–375.

[6] S.B. Sulavik, Bronchocentric granulomatosis and allergic bronchopulmonary aspergillosis, Clin. Chest Med. 9 (1988) 609–621.

[7] J. Languepin, P. Scheinmann, B. Mahut, et al., Bronchial casts in children with cardiopathies: the role of pulmonary lymphatic abnormalities, Pediatr. Pulmonol. 28 (1999) 329–336.

[8] M. Seear, H. Hui, F. Magee, et al., Bronchial casts in children: a proposed classification based on nine cases and a review of the literature, Am. J. Respir. Crit. Care Med. 1 (1997) 364–370.

[9] T.V. Brogan, L.S. Finn, D.J. Pyskaty, et al., Plastic bronchitis in children: a case series and review of the medical literature, Pediatr. Pulmonol. 34 (2002) 482–487.

[10] Gal Neuman, Gideon Koren, Safety of procedural sedation in pregnancy, J. Obstet. Gynaecol. Can. 35 (2) (2013) 168–173.

[11] P.S. Nair, Bronchoscopic cryotherapy in the treatment of plastic bronchitis, CHEST J. 136 (2009) (4_MeetingAbstracts), 23S-c.

[12] T.V. Brogan, L.S. Finn, D.J. Pyskaty Jr., G.J. Redding, D. Ricker, A. Inglis, R.L. Gibson, Plastic bronchitis in children: a case series and review of the medical literature, Pediatr. Pulmonol. 34 (2002) 482–487.